

# An unusual complication of dengue infection

Chien J, Ong A, Low S Y

## ABSTRACT

**We present an unusual complication of dengue infection resulting in postviral phrenic neuropathy and diaphragmatic paralysis in a 34-year-old man. There is a paucity of literature on this condition, with postviral neuropathies previously reported to be associated commonly with herpes zoster, poliovirus, and rarely, West Nile virus and human immunodeficiency virus infections. To our knowledge, this is the first reported case of flavivirus causing isolated postviral phrenic neuropathy and diaphragmatic paralysis.**

**Keywords:** dengue infection, diaphragmatic paralysis, postviral phrenic neuropathy

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## INTRODUCTION

Dengue infection is caused by flavivirus and is the most important arthropod-borne infection in Singapore.<sup>(1)</sup> Most patients recover from dengue infection with minimal or no residual illness.<sup>(2-4)</sup> Dengue infections are usually asymptomatic, but can present with classic dengue fever, dengue haemorrhagic fever or dengue shock syndrome. Some unusual manifestations of dengue infection previously described include liver failure,<sup>(5)</sup> encephalopathy,<sup>(6)</sup> and Guillain-Barré syndrome.<sup>(7)</sup> Post-dengue infection complications are rare and there have been no previous reports on isolated post-dengue infection phrenic neuropathy and diaphragmatic paralysis.

## CASE REPORT

A 34-year-old male smoker with a background history of mild intermittent asthma was admitted with a three-day history of fever and generalised myalgia. His daughter who lives with him had recently been ill with dengue infection. His full blood count showed a total white blood cell count of  $7.7 \times 10^9/L$  and normal differential counts with evidence of haemoconcentration (haematocrit 52.7), and marked thrombocytopenia with platelet count of  $1.0 \times 10^9/L$ . He had a normal chest radiograph on admission (Fig. 1). He was diagnosed with dengue infection as evident by positive serum dengue ribonucleic acid, viral isolation via polymerase



**Fig. 1** Chest radiograph on first presentation is normal.



**Fig. 2** Chest radiograph taken three weeks later shows a right elevated diaphragm.

chain reaction and positive immunoglobulin M serology. He was managed supportively with fluids and transfusion. His illness was complicated by a right pleural effusion, which was treated conservatively and no instrumentation, chest tap or cephalic central venous line was inserted. He was discharged after nine days of hospitalisation.

Department of  
Respiratory and  
Critical Care  
Medicine,  
Singapore General  
Hospital,  
Outram Road,  
Singapore 169608

Chien J MF, MBBS  
Medical Officer

Ong A, MBBS  
House Officer

Low SY, BMBCh,  
MRCP  
Consultant

**Correspondence to:**  
Dr Jaime Chien Mei  
Fong  
Department of  
General Medicine,  
Changi General  
Hospital,  
2 Simei Street 3,  
Singapore 529889  
Tel: (65) 6788 8833  
Fax: (65) 6781 6202  
Email: jaimechien@  
yahoo.com.sg

Three weeks later, he was admitted for the complaints of dyspnoea and cough of three days' duration. Clinical examination revealed normal vital signs and decreased air entry on the right with dullness to percussion. Chest radiograph (Fig. 2) revealed an elevated right hemidiaphragm with no pleural effusion as evident by no layering on a right lateral decubitus film. Computed tomography (CT) of the thorax did not reveal any lung parenchymal, mediastinal abnormalities or liver lesions. He subsequently underwent a nerve conduction study of the phrenic nerves, which revealed decreased conduction amplitudes bilaterally, affecting the right more than the left diaphragm. Electromyography studies to look out for myasthenia gravis were performed with normal results. Fluoroscopy sniff test showed an absence of movement of the right hemidiaphragm with respiration. His lung function test revealed a sitting forced vital capacity of 2.11 L and supine 1.2 L (difference of 43%). He was managed expectantly with gradual symptomatic improvement and was discharged stable on the fifth day of hospitalisation.

## DISCUSSION

Dengue infection is endemic in Singapore and is the most important arthropod-borne disease in Singapore.<sup>(1)</sup> It is transmitted through the bite of an infected day-biting mosquito, most commonly *Aedes aegypti*. Most infected human hosts are asymptomatic. Classic dengue fever develops in 15%–60% of patients<sup>(2,4)</sup> with symptoms of fever, headaches, retro-orbital pain, myalgia, petechiae rash with “islands of sparing”. Dengue haemorrhagic fever is characterised by the World Health Organisation with four cardinal features; viz. fever for 2–7 days; evidence of plasma leakage (haemoconcentration with haematocrit elevated > 20% baseline, pleural effusion, and ascites); haemorrhagic tendencies or spontaneous bleeding; and marked thrombocytopenia (platelet count < 100,000/mm<sup>3</sup>).<sup>(8)</sup> An estimate of 20%–30% of patients with dengue haemorrhagic fever develop dengue shock syndrome.<sup>(9)</sup> Other uncommon manifestations of dengue infection reported include fulminant liver failure,<sup>(5)</sup> neurological disturbances of encephalopathy,<sup>(6)</sup> Guillain-Barré syndrome,<sup>(7)</sup> transverse myelitis and polyradiculoneuritis.<sup>(7,10-13)</sup>

Our patient had dengue infection, which was later complicated with postviral phrenic neuropathy resulting in bilateral diaphragmatic paralysis, affecting the right side more than the left. This was suggested by a positive fluoroscopy sniff test, a more than 20% difference in vital capacity between sitting and supine lung function testing and compatible nerve conduction studies. Electromyography did not suggest a myopathic aetiology and CT of the thorax did not reveal possible pathologies

that could result in phrenic nerve compression. He was managed expectantly and his symptoms resolved by his second outpatient review one month later.

Postviral phrenic neuropathy resulting in diaphragmatic paralysis are rare occurrences and have been previously described in case reports in association with poliovirus infection,<sup>(7)</sup> herpes zoster infection,<sup>(14)</sup> West Nile virus,<sup>(15)</sup> and human immunodeficiency virus.<sup>(16)</sup> We report an unusual complication of dengue infection resulting in postviral phrenic neuropathy causing diaphragmatic paralysis. The course of postviral phrenic neuropathy remains undefined in the current literature due to a paucity of reports. Patients with postviral phrenic neuropathy and diaphragmatic paralysis were previously reported to run a benign course or sustain a poor outcome with severe life-threatening respiratory failure needing mechanical ventilation.<sup>(14,15)</sup> Polyradiculoneuritis is a well-described complication of dengue infection, especially in the form of Guillain-Barré syndrome.<sup>(7)</sup> Isolated phrenic neuropathy, however, has not been previously described in the polyradiculoneuritis associated with post-dengue infection. This patient had an isolated neuropathy of the phrenic nerves, and his clinical course did not fit into that of Guillain-Barré syndrome. This is the first reported isolated case of phrenic neuropathy in a patient who had had dengue infection. This case illustrates that post-dengue infected patients may develop phrenic neuropathy as an isolated neurological complication. In a tropical country with endemic dengue infections, post-dengue infection phrenic neuropathy may be considered among the causes of diaphragmatic paralysis.

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